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CASE REPORT

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Developmental spectrum of segmental odontomaxillary dysplasia: clinical and radiographic features across different dentition stages

Hwajung Yong[1,†], Suji Lee[1,†], Jaesik Lee[1,*]

[1]Department of Pediatric Dentistry, School of Dentistry, Kyungpook National University, 41940 Daegu, Republic of Korea

***Correspondence**
leejs@knu.ac.kr
(Jaesik Lee)

[†] These authors contributed equally.

Abstract

Background: Segmental odontomaxillary dysplasia (SOD) is a rare developmental condition characterized by unilateral maxillary involvement with dental and osseous abnormalities. Although several case reports have described individual features of SOD, clinical and radiographic findings across different stages of dental development remain limited. This report describes the clinical and radiographic findings of SOD observed at different stages of dentition. **Cases**: Three pediatric patients with SOD were evaluated at different stages of dentition, including the primary dentition and late mixed dentition, with emphasis on imaging findings. Clinical examinations and radiographic evaluations, including panoramic radiography and cone-beam computed tomography, were performed. **Conclusions**: Clinical and radiographic findings in two patients examined during the primary dentition stage were primarily limited to the affected maxillary segment and primary dentition, including abnormal primary tooth morphology and displacement of developing permanent tooth germs. In contrast, the patient evaluated during the late mixed dentition stage demonstrated additional findings involving the permanent dentition, including ankylosis, congenitally missing premolars, and delayed eruption of the permanent canine. Follow-up radiographs showed partial eruption of the affected canine after extraction of obstructive primary teeth.

Keywords

Segmental odontomaxillary dysplasia; Dental development; Mixed dentition; Cone-beam computed tomography

1. Introduction

Segmental odontomaxillary dysplasia (SOD) is a rare developmental disorder of unknown etiology that causes unilateral alteration of the maxilla, associated with abnormal growth and maturation of bone, absence of one or both premolars and delayed tooth eruption on the affected side, and alteration of adjacent gingival tissue with or without facial cutaneous lesions [1–3]. It was first described by Danforth *et al.* [4] in 1990, characterized by a unilateral dysplasia involving the maxillary alveolus and the associated dentition. Although cases have been documented in both sexes, previous reports suggest a slight male predominance [5].

Clinically, SOD typically manifests as unilateral buccolingual expansion of the posterior maxilla accompanied by gingival or alveolar enlargement. Affected patients often exhibit dental spacing or asymmetry confined to the involved segment. Radiographically, characteristic findings include a coarse and thickened trabecular bone pattern, morphological anomalies of the primary posterior teeth, and displacement or agenesis of permanent successors. The frequent absence of premolar tooth germs, delayed eruption patterns, and structural variations in erupted or unerupted teeth further support the diagnosis of SOD [6].

Despite being recognized as a rare developmental condition, SOD has not been comprehensively characterized in terms of its clinical features, complicating its consistent description in the dental literature. The clinical presentation appears to be associated with patient age and the stage of dental development, which may pose considerations in early diagnosis. SOD in the primary dentition may be suspected based on atypical crown or root morphology of the maxillary primary canines and molars, whereas patients in the mixed dentition often present with displacement or congenital absence of permanent teeth. At later stages, radiographic examination may demonstrate skeletal changes such as unilateral maxillary enlargement. These stage-dependent differences highlight that the clinical focus in patients with SOD may vary according to dental developmental stage, particularly with respect to diagnostic emphasis and follow-up considerations.

Although SOD has been described in several case reports, clinicians may still encounter difficulties in recognizing and interpreting its dental features at different stages of development. This is partly because the existing literature has

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not comprehensively illustrated the spectrum of clinical and radiographic findings across developmental stages. Therefore, the present case report compares three patients evaluated at different stages of dental development to better characterize stage-specific manifestations of SOD and discuss their diagnostic implications.

2. Case presentations

Clinical and radiographic examinations, including panoramic radiography and cone-beam computed tomography (CBCT), were performed in all patients. CBCT images were acquired using the HDXWILL EcoX-ai (HDXWILL, Seoul, Korea) with the following imaging parameters: 85 kVp, 6 mA, 24-second exposure time, 16 × 9 cm field of view, and a voxel size of 0.20 mm. Image datasets were reviewed using the hospital's INFINITT Picture Archiving and Communication System (PACS) software (version G3 30116 BN2; Infinitt Co., Ltd., Seoul, Korea).

2.1 Case 1

A 4-year 6-month-old boy first visited our institution on 23 August 2019 with a chief complaint of morphologic abnormalities of the left maxillary primary molars. Intraoral examination demonstrated abnormal crown morphology of the maxillary left primary canine, first molar, and second molar (#63–65). These teeth showed incomplete eruption and appeared partially embedded within the gingival tissues, resulting in an irregular occlusal contour. In addition, occlusal wear of the affected primary teeth was noted. The posterior left maxillary segment exhibited unilateral buccolingual enlargement of the gingiva and alveolar process (Fig. 1).

Periapical and panoramic radiographs revealed distinct morphologic alterations of the maxillary left primary canine (#63), first molar, and second molar (#64–65), including divergent roots and enlarged pulp chambers (Fig. 2). Notably, the maxillary left primary canine (#63), which typically presents with a single root, displayed an atypical root configuration suggestive of bifurcation (Fig. 2). CBCT findings further presented widened root divergence and enlarged pulp morphology (Fig. 3). The affected quadrant exhibited congenital absence of the maxillary left second premolar (#25), with displacement of the developing permanent tooth germs, including the maxillary left lateral incisor (#22) and first premolar (#24) (Fig. 2). Axial and coronal CBCT images illustrated an altered trabecular bone pattern and buccolingual expansion of the alveolar bone extending from the maxillary left primary canine (#63) to the posterior maxillary segment (Fig. 3). The most recent follow-up examination was performed on 30 March 2026.

2.2 Case 2

A 5-year 5-month-old boy first visited our institution on 14 July 2025, with a chief complaint of abnormal morphology of the maxillary right primary first molar. Clinical examination revealed unilateral enlargement of the posterior right maxillary alveolar process, with buccolingual expansion of the gingiva and underlying bone. The maxillary right primary first molar (#54) exhibited an irregular crown contour with poorly defined cusps and an atypical occlusal morphology. A distinct spacing was noted between the maxillary right primary canine and first molar (#53–54), suggesting localized developmental disturbance on the affected side (Fig. 4).

Panoramic radiography revealed morphologic abnormalities of the maxillary right primary canine, first molar, and second molar (#53–55), including enlarged pulp chambers and divergent root morphology of the primary molars (Fig. 5). In addition, a widened interdental space between the maxillary right primary canine and first molar (#53–54) was evident. CBCT demonstrated buccolingual expansion of the right posterior maxillary alveolar process from #53 to the second primary molar region, with an irregular and coarse trabecular pattern within the involved segment (Fig. 6). Displacement of the developing permanent tooth germs was noted, involving the maxillary right lateral incisor (#12) and first premolar (#14) (Figs. 5,6). The most recent follow-up examination was performed on 17 April 2026.

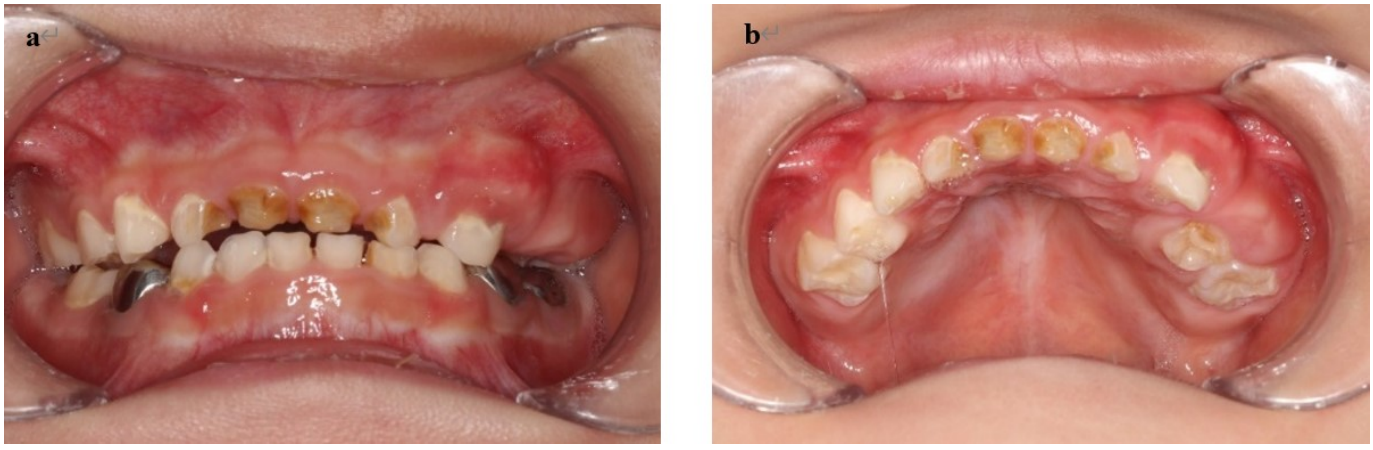


FIGURE 1. Intraoral images of Case 1. (a) Frontal intraoral image showing unilateral enlargement of the affected left maxillary posterior segment. (b) Maxillary occlusal view showing morphological abnormalities of the maxillary left primary teeth.



FIGURE 2. Panoramic radiograph of Case 1 showing morphological abnormalities of the maxillary left primary teeth and displacement of the developing permanent teeth.

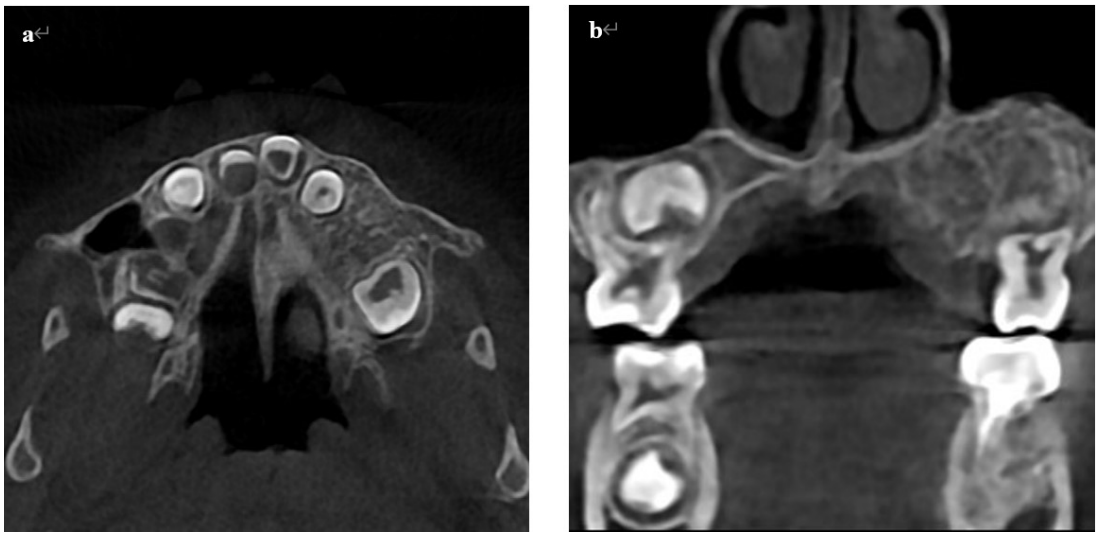


FIGURE 3. Cone-beam computed tomography (CBCT) images of Case 1. (a,b) Axial and coronal CBCT images showing buccolingual expansion of the affected left posterior maxilla.

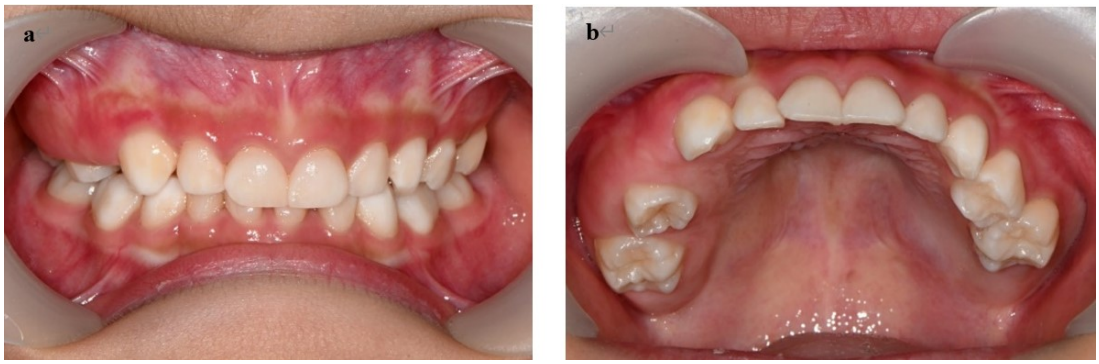


FIGURE 4. Intraoral images of Case 2. (a) Frontal intraoral image of the affected right maxillary posterior segment. (b) Maxillary occlusal view showing the affected right maxillary primary dentition.



FIGURE 5. Panoramic radiograph of Case 2 showing the affected right maxillary primary dentition and developing permanent teeth.

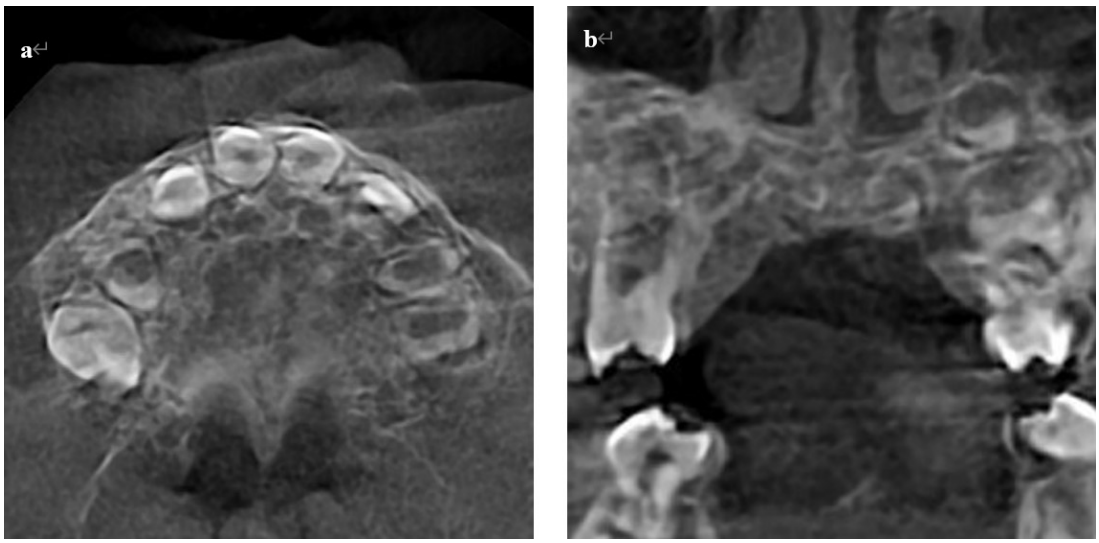


FIGURE 6. Cone-beam computed tomography (CBCT) images of Case 2. (a,b) Axial and coronal CBCT images of the affected right posterior maxilla.

2.3 Case 3

An 11-year 5-month-old boy first visited our institution on 14 March 2025, with a chief complaint of congenital absence of the left maxillary premolars. Clinical and radiographic evaluation revealed multiple alterations involving both the primary and permanent dentitions. Intraoral examination demonstrated morphologic irregularities of the maxillary left primary first and second molars (#64–65), with the maxillary left first molar (#64) exhibiting ankylosis. The posterior left maxillary segment displayed unilateral buccolingual expansion of the alveolar process, consistent with localized enlargement of the affected area.

Panoramic radiography provided further detail regarding these findings. Unlike Cases 1 and 2, which were evaluated during the primary dentition stage, this patient was in the late mixed dentition stage. The maxillary left first and second primary molars (#64–65) showed distinct crown and root structural variations, and ankylosis of the maxillary left first primary molar (#64) was evident (Fig. 7). CBCT images further demonstrated morphological abnormalities of the maxillary left first and second permanent molars (#26–27) and abnormal pulp and root canal morphology, including enlarged and irregular pulp chambers with limited canal differentiation (Fig. 8).

The maxillary left canine (#23) displayed delayed eruption, whereas the corresponding teeth on the unaffected side exhibited normal eruption. The maxillary left first and second premolars (#24–25) were congenitally missing, consistent with the patient's chief complaint. Axial and coronal CBCT views demonstrated an irregular and coarse trabecular bone pattern



FIGURE 7. Panoramic radiograph of Case 3 showing the affected left maxillary dentition with delayed eruption of the permanent canine and congenitally missing premolars.

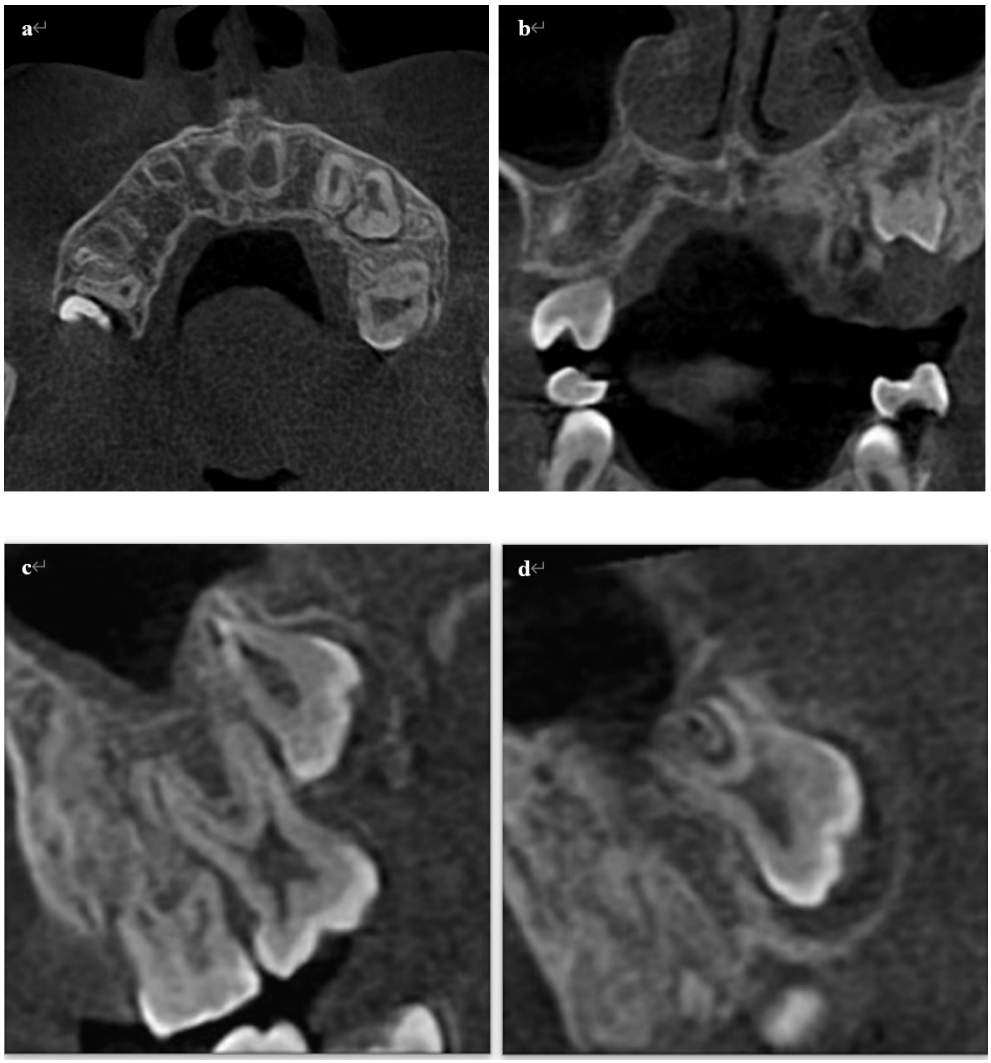


FIGURE 8. Cone-beam computed tomography (CBCT) images of Case 3. (a,b) Axial and coronal CBCT images show expansion of the left maxillary alveolar process. (c) Sagittal CBCT image of the maxillary left first permanent molar (#26). (d) Sagittal CBCT image of the maxillary left second permanent molar (#27).

extending from the region of the maxillary left canine (#23), posteriorly throughout the affected maxillary segment, with pronounced buccolingual expansion of the alveolar bone compared with the contralateral side (Fig. 8).

Given the delayed eruption of #23 and ankylosis of #64, extraction of the maxillary left primary canine (#63) and the ankylosed maxillary left first primary molar (#64) was planned. Intraoral images and a panoramic radiograph obtained 1 week after extraction are shown in Fig. 9, and the 4-month follow-up images are presented in Fig. 10. The most recent follow-up examination was performed on 05 December 2025.

Table 1 summarizes the dental and maxillary findings observed in the three cases, based on the clinical and radiographic features described above.

3. Discussion

Patients in the primary dentition stage (Cases 1 and 2) exhibited abnormalities of primary teeth, localized gingival enlargement, and characteristic trabecular bone patterns, whereas the patient in the late mixed dentition stage (Case 3) demonstrated additional involvement of permanent teeth and eruption disturbances (Table 1).

SOD has been described in the literature as a rare, localized developmental disorder of the maxilla, characterized by unilateral maxillary enlargement, morphological abnormalities of the primary molars, displacement or absence of premolars, and a coarse trabecular bone pattern. These features were initially summarized by Danforth *et al.* [4], who emphasized the segmental distribution of the osseous and dental changes. Subsequent case reports by Becktor *et al.* [7], Packota *et al.* [8], and González-Arriagada *et al.* [9] further broadened the range of reported observations, describing associated features, such as gingival enlargement, altered eruption patterns, and

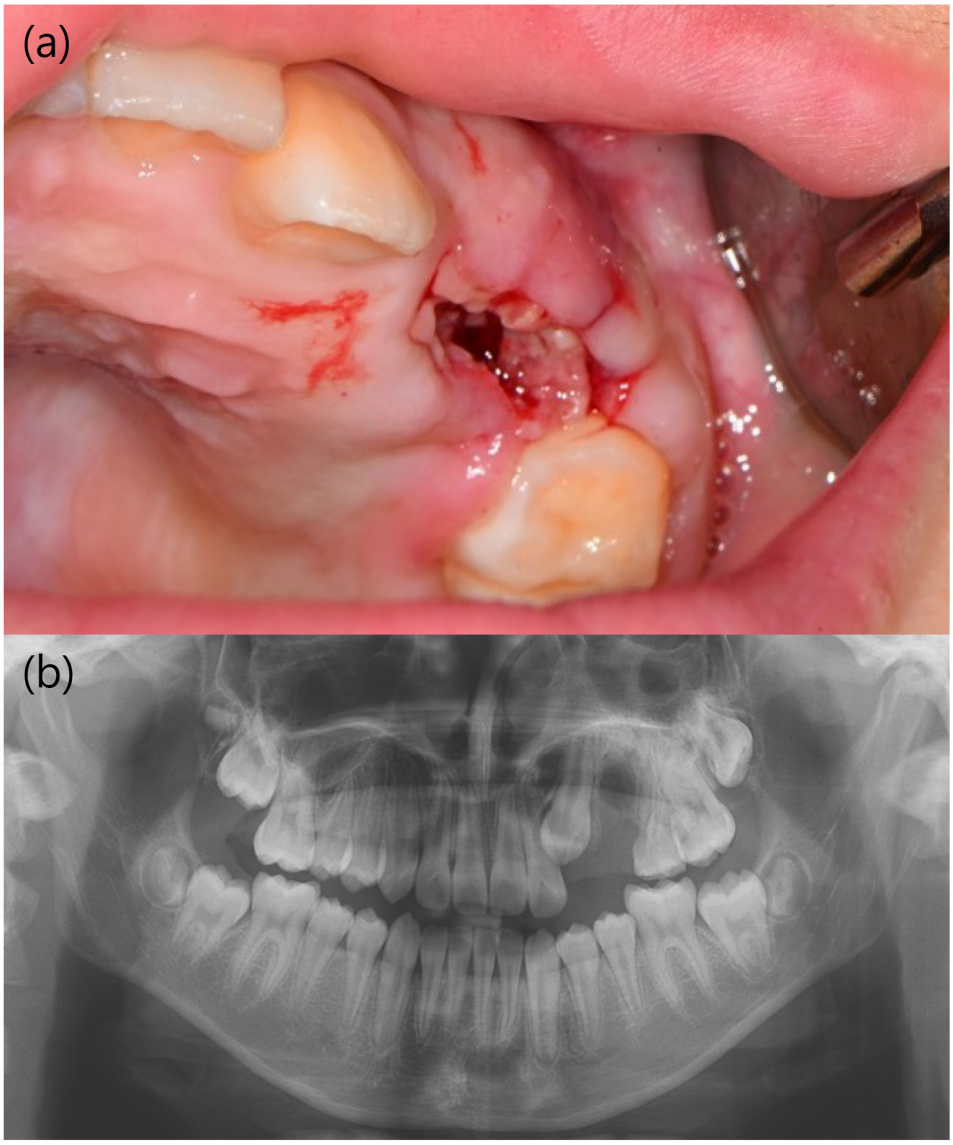


FIGURE 9. Intraoral image and panoramic radiograph of Case 3 obtained 1 week after extraction. (a) Intraoral image of the extraction site of the maxillary left primary canine (#63) and primary first molar (#64). (b) Panoramic radiograph obtained after the extraction of the maxillary left primary canine (#63) and primary first molar (#64).

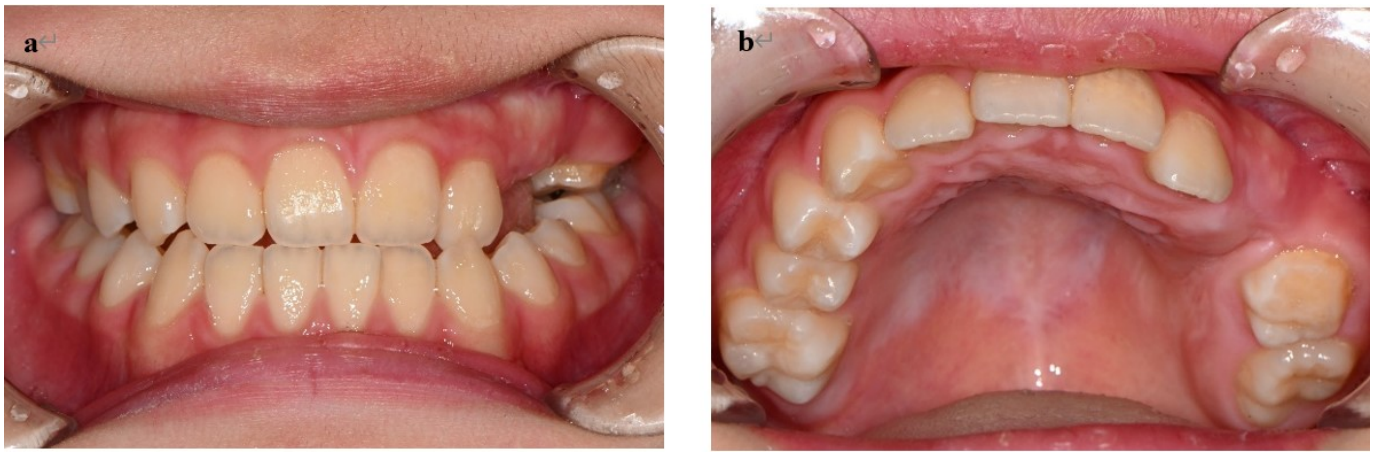


FIGURE 10. Intraoral images of Case 3 obtained 4 months after extraction. (a) Frontal intraoral image of the extraction site of the maxillary left primary canine (#63) and the first primary molar (#64). (b) Maxillary occlusal view obtained 4 months after extraction.

TABLE 1. Distribution of dental and maxillary features observed in the three SOD cases.

	Case 1	Case 2	Case 3
Dental developmental stage	Primary dentition	Primary dentition	Late mixed dentition
Unilateral maxillary involvement	Posterior left maxilla	Posterior right maxilla	Posterior left maxilla
Buccolingual gingival enlargement	Posterior left maxilla	Posterior right maxilla	Posterior left maxilla
Buccolingual alveolar bone expansion	Primary canine to second primary molar region (left)	Primary canine to second primary molar region (right)	Canine to second permanent molar region (left)
Abnormal morphology of teeth	Primary canine and primary molars (left)	Primary canine and primary molars (right)	Primary and permanent molars (left)
Coarse trabecular bone pattern (CBCT)	From the maxillary left primary canine posteriorly	From the maxillary right primary canine posteriorly	From the maxillary left canine posteriorly
Displacement of permanent tooth germs	Maxillary left lateral incisor and first premolar	Maxillary right lateral incisor and first premolar	Maxillary left canine
Congenital absence of permanent premolars	Maxillary left second premolar	Not observed	Maxillary left first and second premolars
Delayed eruption of permanent teeth	Not observed	Not observed	Maxillary left canine
Ankylosis	Not observed	Not observed	Maxillary left primary first molar
Involvement of permanent molars	Not observed	Not observed	Maxillary left permanent first and second molars

variable involvement of canines or permanent molars. Furthermore, previous reports have suggested that the clinical and radiographic appearance of SOD may differ according to patient age and the stage of dental development, possibly influencing clinical recognition and differentiation from other developmental or fibro-osseous conditions of the maxilla [1, 9]. However, comparative descriptions across different developmental stages remain limited, making it difficult to appreciate the spectrum of SOD manifestations throughout dental development.

The present cases share several characteristics described in earlier reports. The affected segments in Cases 1 and 2, both in the primary dentition stage, displayed morphological alterations of the primary teeth, including enlarged pulp chambers, splayed roots, and atypical crown morphology extending from the canines to the molars. Similar features were previously noted by Armstrong *et al.* [10] and Becktor *et al.* [7], who proposed that impaired odontogenesis within the dysplastic region may contribute to these dental variations. Cone-beam computed tomography (CBCT) further revealed a coarse and irregular trabecular bone pattern, a finding frequently associated with dysplastic maxillary bone in SOD [7]. These CBCT findings not only confirmed the suspected dysplastic bone architecture, but also facilitated the assessment of the

developmental status of permanent tooth germs and treatment planning. Packota *et al*. [8] reported that displacement of successor teeth may precede later eruption disturbances or agenesis.

Case 3, representing a patient in the late mixed dentition stage, provides additional information regarding the manifestation of SOD beyond the primary dentition. Ankylosis of the primary first molar was noted, together with congenital absence of the corresponding permanent successors, a pattern that has been described in previous reports and may be associated with disturbances in the formation or mineralization of successor tooth germs. In addition, permanent molars within the affected segment exhibited structural variations involving both external morphology and internal pulp configuration. These observations suggest that SOD may be associated with additional dental structural changes in later stages of dental development, beyond those typically described in earlier stages [7, 9]. Although involvement of canines and permanent molars has been infrequently reported in the literature, these findings indicate that such features can be present in patients examined at later stages of dental development [8, 9]. When interpreted together with Cases 1 and 2, these observations provide a comparative perspective on the clinical manifestations of SOD across different stages of dental development.

Short-term follow-up in this case provided further insights into the clinical course of these findings. These follow-up findings possibly reflect changes in the local eruptive environment following removal of ankylosed or obstructive primary teeth, rather than resolution of the underlying dysplastic process. Alterations in maxillary bone architecture associated with SOD may be associated with changes in the local eruptive environment and could affect the eruption timing of the adjacent permanent teeth [11]. In this case, delayed eruption of the permanent canine may be related to a combination of local mechanical factors, such as ankylosis of the primary molar, and coexisting alterations in the alveolar bone. Because involvement of canines and permanent molars has been only sporadically reported in the literature, this case highlights understudied aspects of SOD and underscores the significance of longitudinal monitoring. Further investigation is warranted to clarify how SOD affects eruption timing, root development, and occlusal relationships throughout dentition.

The clinical characteristics identified across these cases illustrate several considerations in providing dental care for children affected by SOD. Although pulpotomy was indicated in Case 1 due to deep caries, gingival and alveolar enlargement, together with atypical tooth morphology, interfered with the stable placement of a rubber dam clamp. Difficulty in securing the clamp during the procedure caused a near-aspiration event, highlighting an additional clinical risk associated with inadequate isolation. This is clinically relevant, as effective isolation is fundamental for pulp therapy and restorative procedures, particularly in pediatric patients. Furthermore, abnormal root morphology, such as taurodontism-like pulp chambers, divergent roots, or irregular canal morphology, complicates conventional endodontic access and instrumentation. These variations may increase the risk of procedural errors, incomplete pulp removal, or inadequate obturation. These challenges highlight the significance of implementing preventive strategies at an early age to minimize the need for complex restorative or endodontic procedures. Preventive approaches may include topical fluoride application, sealants, early caries risk assessment, and frequent recall visits [9, 11, 12]. In the present case, the abnormal gingival and dental morphology also caused difficulty in securing the rubber dam clamp. Therefore, clinicians should anticipate potential difficulties with isolation devices and consider alternative isolation methods or modified techniques when treating patients with SOD.

Case 3 also demonstrated important considerations regarding post-extraction healing and surgical planning in SOD. The affected tooth exhibited ankylosis, a condition commonly associated with altered bone remodeling dynamics. Radiographic imaging confirmed a thickened and coarse trabecular bone architecture—a hallmark of SOD, possibly indicating underlying dysplastic bone formation [11, 13]. A clinical examination performed 1 week after extraction revealed delayed healing of both the alveolar bone and the surrounding soft tissues. Although a direct causal relationship between coarse trabeculation and impaired healing has not been conclusively established, the existing literature suggests that such bone morphology may reflect atypical mineralization, reduced vascularity, or altered turnover rates. These factors could contribute to slower or less predictable healing outcomes. These considerations are particularly relevant when planning future interventions, such as orthodontic tooth movement, implant placement, or alveolar osteoplasty. Careful assessment of bone quality and density in patients approaching skeletal maturity is essential to determine the feasibility and timing of surgical correction. Moreover, clinicians should provide detailed preoperative and postoperative counseling to patients and caregivers, setting appropriate expectations regarding healing time, potential complications, and long-term management strategies. Unlike previous reports that primarily described individual presentations of SOD, the present case series enables comparison of clinical and radiographic findings observed at different stages of dental development. Although these cases do not represent longitudinal observations of the same patient, they illustrate the range of manifestations that may be encountered throughout childhood and provide practical information that may facilitate earlier recognition and clinical assessment of this rare condition.

This study has several limitations. First, the findings are based on only three cases, and the descriptive nature of this case report should be considered when interpreting the findings. Second, histopathological confirmation was not available. Finally, the relatively short follow-up period limited evaluation of long-term clinical and radiographic findings. Further accumulation of well-documented cases with longer follow-up is needed to better characterize the clinical and radiographic features of SOD.

4. Conclusions

This case report presents three patients with SOD who were evaluated at different stages of dental development. Differences in clinical and radiographic features, including tooth morphology, eruption patterns, and maxillary bone involvement, were observed among the three patients. Recognition

of these stage-related features may assist clinicians in diagnosis, follow-up planning, and dental management of pediatric patients with SOD. Further case-based observations may help to better characterize the clinical and radiographic features of SOD across different stages of dental development.

ABBREVIATIONS

SOD, segmental odontomaxillary dysplasia; CT, computed tomography; CBCT, Cone-beam computed tomography; PACS, Picture Archiving and Communication System.

AVAILABILITY OF DATA AND MATERIALS

All data generated or analyzed during this study are included in this published article.

AUTHOR CONTRIBUTIONS

HJY—designed the research study. HJY and SJL—collected the clinical data and wrote the manuscript. JSL—critically revised the manuscript. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

The Kyungpook National University Dental Hospital Institutional Review Board approved this study (protocol no. KNUDH-2026-03-02-00). Written informed consent was obtained from the patients' parents or legal guardians, and all patient information was anonymized before analysis.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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